Penile Crohn's disease: a case report

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Abstract

Metastatic Crohn's disease is a rare inflammatory condition characterised by cutaneous granulomatous lesions separated from the affected bowel by normal intact skin. Involvement of the genitalia in Crohn's disease is rare and consists of ulcerated lesions in almost all of the cases reported in the literature. We describe a case of penile involvement in a 27 year old man with a 5 year history of Crohn's disease. Should genital involvement precede the bowel disease, patients may consult the sexually transmitted disease service for this problem and the dermatovenereologists may be the first to formulate the diagnosis.

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Introduction

Non caseating epithelioid granulomas are the pathological features of skin involvement in several diseases such as systemic sarcoidosis, foreign body and necrobiotic granulomas, and some mycobacterial and fungal infections; very rarely these pathological features may represent cutaneous Crohn's disease (CD).[1]

The onset of cutaneous or mucosal lesions of CD most frequently follow involvement of the bowel.[2] It may occur in sites adjacent to bowel such as around the anus, or at colostomy or ileostomy sites or in sites unrelated to the bowel: these cases constitute so-called metastatic Crohn's disease (MCD),[3] a rare inflammatory condition presenting with ulcers, papules, nodules and plaques. Involvement of genitalia by MCD is rare.

Case report

A 27 year old man presented to our sexually transmitted disease service with a 14 day history of painless penile ulceration. On clinical examination diffuse erythema and swelling of the glans and preputial skin were evident. An oval ulcer of 1 cm at its greatest diameter with an indurated base was localised at the base of the glans and on the inner preputial skin. Three minor oval lesions were found near the principal ulcer.

There was no significant inguinal lymphadenopathy and the patient did not have any other genitourinary symptoms. A cytological examination of the ulcers showed a mixture of inflammatory cells and dark field microscopy did not reveal *Treponema pallidum*.

The patient had been diagnosed as having Crohn's colitis five years prior to our examination, following several episodes of abdominal pain and bloody diarrhoea. The diagnosis was confirmed by radiology, endoscopic examination and biopsy of the gastrointestinal tract. At first treatment with methylprednisolone 20mg/day associated with sulphasalazine had been started. Sulphasalazine, however, had been interrupted because of a severe diffuse acute urticaria. In 1991 the patient had a colectomy and ileostomy because medical therapy had produced only slight improvement. Macroscopically the colon had a cobblestone appearance with multiple ulceration and pseudopolyps.

Two months after surgery the patient had experienced a relapse of the gastrointestinal Crohn's disease and was treated with cyclosporin A (300 mg/day) with mild improvement. After four months this was stopped so that at the time of presentation to our department the patient was not receiving any therapy.

Haematological analysis showed a white cell count of 16·7/mm^3 with neutrophilia and

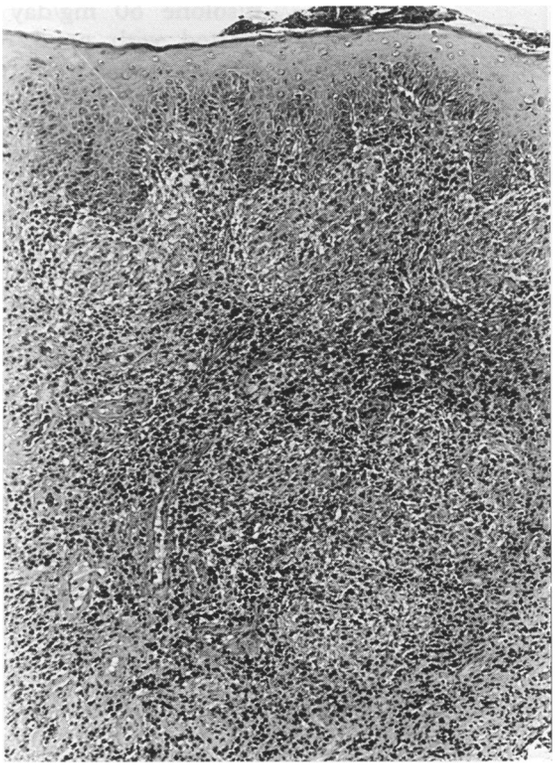

Figure 1 Pathological aspect of the border of the penile ulcer: a non caseating granulomatous infiltrate involves the full thickness of the tonaca propria. (H and E 80×)

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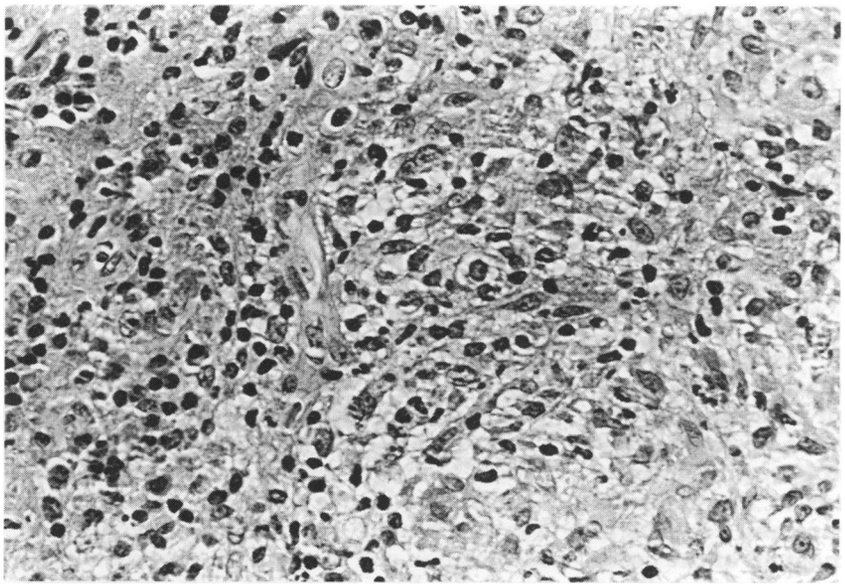

Figure 2 High power view of the granuloma that is constituted of epithelioid cells surrounded by lymphocytes and plasma cells. (H and E 200×)

an increased ESR (20 mm in one hour). Results of chemistry studies were within normal ranges. A biopsy specimen from the penile ulcer showed a non caseating granulomatous inflammation involving the full thickness of the tunica propria. Granulomas contained epitheliod cells and rare Langerhans giant cells surrounded by a dense infiltrate formed of lymphocytes and many plasma cells (fig 2). Polarising microscopy was negative. Fungal and acid-fast stains were negative and cultures for mycobacteria, fungi and bacteria from the biopsy specimen were negative. This pathological picture was considered consistent with cutaneous Crohn's disease.

Treatment with systemic methylprednisolone 60 mg/day and topical CyA was started with good response. The systemic steroid dosage was progressively reduced after two weeks of therapy. One month later the ulcer had completely healed. At six months follow-up he remained well.

Discussion

MCD was first described in 1965 by Parks, Morson and Pegum.[4] It is a rare condition characterised by cutaneous granulomatous lesions separated from the affected bowel by normal intact skin. The cause of MCD is still unknown. Some authors have suggested two possible pathogenetic mechanisms: a granulomatous response to antigens carried to the skin (granulomatous perivasculitis)[5]; or a variety of granulomatous vasculitis,[6 7] a vascular immune reaction mediated by sensitised T lymphocytes reacting to circulating antigen.[8] The clinical appearance of MCD is quite variable: nodules, plaques or ulcers, most commonly localised on the extremities or in intertriginous areas, although any site on the body may be involved.[9] Genital involvement is, however, exceptional but has been described in both men and women.[4 10]

In 1970, studying 207 patients with Crohn's disease, Montain[1] reported four cases of MCD including one case of penile involvement. Schum[3] in 1990 reviewed the literature and reported four further cases of MCD of genitalia.[3 11 12] In almost all cases penile lesions were ulcers and were not associated with widespread MCD.[3] The diagnosis of MCD should be obvious if the patient has associated intestinal manifestations, but sometimes the ulcerated genital lesions may precede the onset of gastrointestinal symptoms. In these cases the differential diagnosis include sexually transmitted genital ulcer diseases such as chancroid, lymphogranuloma venereum, granuloma inguinale and herpes genitalis.[11 13]

The pathological features of MCD include epidermal ulceration, dermal or subdermal noncaseating granulomas containing epithelioid cells and a variable number of Langerhans/giant cells, dermal abscesses and a plasma cell and lymphocytic infiltrate. Although cutaneous Crohn's disease does not have a pathognomic pathology, the above features can be highly suggestive of this diagnosis if accompanied by general symptoms or where there is a history of previous bowel surgery.[3]

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